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SURVEY (SYSTEMATIC REVIEW)

Subungual Exostosis of the Toes: A Systematic Review

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Abstract

Background   Subungual exostosis is a relatively common benign bone tumor that occurs in the distal phalanges of the toes and can be a source of pain and nail deformity. There is controversy about the treatment of these lesions and there are few studies that have synthesized what is known and provided meaningful information on treatment.

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Questions/purposes   We performed a systematic review to address the following questions: (1) What is the best surgical approach for excising these lesions? (2) What is the age range, sex distribution, and presenting symptoms of subungual exostoses and which toe is most frequently affected? (3) What complications arise from treatment?
Methods   Two authors independently searched multiple databases (Medline, 1950–May 2013; Cochrane EBM database, and EMBASE, 1980–May 2013 provided by OVID; ACP Journal Club, 2003–May 2013; CINAHL by EBSCO, 1937–May 2013; and PubMed by NLM, 1940–May 2013), and key words were chosen to achieve a broad search strategy. We included studies on the management of toe exostoses with > 10 cases and we excluded studies that reported on upper extremity exostoses or osteochondromas. Demographic and treatment data were collected from each article by two independent authors and collated. A total of 124 abstracts were screened, and 116 articles were reviewed in full, of which 13 met the inclusion criteria.
Results   Complete marginal excision through a fish mouth incision protecting the nail led to a recurrence rate of 4% and satisfactory clinical results, defined as no requirement for postoperative intervention and a satisfactory clinical appearance in 73%. Most studies provided incomplete descriptions of specific surgical techniques used. Fifty-five percent of the patients were younger than 18 years of age. A history of toe trauma before diagnosis was present in approximately 30% of the cases. Delayed diagnosis occurred in approximately 10% of the cases and onychodystrophy occurred in more than 10%.
Conclusions   There is weak evidence to guide management of subungual exostosis. Adequate wound management postexcision aiming to minimize disruption to the nail bed and matrix may prevent onychodystrophy, which is a common complication of treatment.

Introduction

Subungual exostosis is a relatively uncommon, osteocartilaginous tumor that affects the distal phalanx of the toes or fingers [2, 5, 12, 24] (Fig. 1). This benign lesion has also been reported as part of multiple hereditary exostoses [1, 24, 28, 32]. Dupuytren described in 1817 a bony growth of the distal phalanx of the great toe that became known as "Dupuytren's exostosis." The exact pathogenesis of subungual exostosis is unclear, although trauma, infection, tumor, hereditary abnormality, or activation of a cartilaginous cyst all have been suggested as possible etiologies of subungual exostosis. Most consider it to be a reactive metaplasia resulting from microtrauma [3, 33, 35]; however, there is no conclusive evidence to support a single pathogenesis [14, 32, 34]. There is debate whether subungual osteochondroma is the same clinical entity as subungual exostosis. Histologically, the cartilaginous cap of exostoses is made of fibrocartilage, whereas in osteochondromas, it is hyaline cartilage and is confluent with the underlying trabecular and cortical bone [16, 25] (Fig. 2). In exostoses, bone is formed directly from fibrous tissue, whereas in conventional osteochondromas, it is derived from enchondral ossification [12, 16, 24]. The translocation t(X;6)(q22;q13-14) has been reproducibly linked to subungual exostosis [7, 33], implying it is a true neoplasm instead of being a reactive process in response to trauma. Other lesions of the bone surface such as bizarre parosteal osteochondromatous proliferation (BPOP or Nora lesion), once thought to be a related proliferative process to subungual exostoses, have unique chromosomal rearrangements and represent a distinct molecular pathogenesis [33].

The most common clinical presentation is that of several months of pain, erythema, and deformity of the nail bed [8, 12, 33, 38], which cause a deleterious impact on health-related quality of life [8, 36]. Examination usually reveals a firm, fixed nodule with a hyperkeratotic smooth surface at the distal end of the nail plate. Because misdiagnosis and delayed diagnosis of this lesion are common, appropriate treatment often is not rendered [31, 37]. Radiographs show a pedunculated

Fig. 1A–D Clinical photographs and radiographs of the same subungual exostosis in the right hallux of a 15-year-old girl presenting with pain and an enlarging mass for 3 months. (**A**) Dorsal and (**B**) medial clinical photographs and (**C**) posterior-anterior and (**D**) lateral radiographs of the same lesion. Reproduced with permission from *BMJ Case Reports* [6].
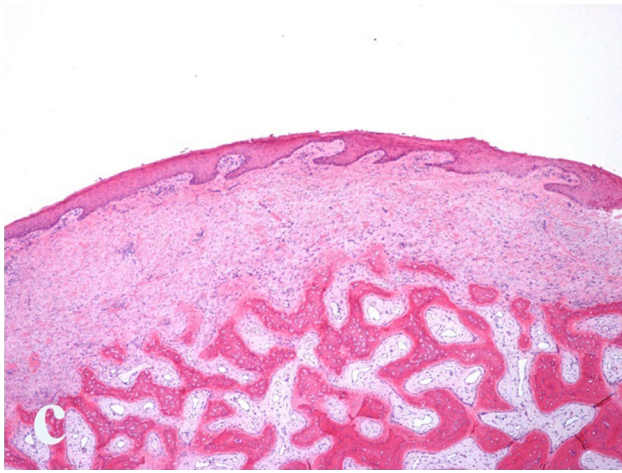
Fig. 2 Light microscopic image of a histological specimen taken intraoperatively from the great toe of a 15-year-old girl. It shows dermal tissue with normal trabecular bone and fibrocartilaginous overgrowth, consistent with subungual exostosis. Reproduced with permission from *BMJ Case Reports* [6].

radioopaque mass on the dorsomedial surface of the distal phalanx. The role of nonoperative management is limited because the condition is generally progressive. The mainstay of treatment has been marginal surgical excision of the exostosis, which typically alleviates the symptoms [25–27]. However, surgical techniques have varied [2, 11, 17, 33], and the most recent literature review on this topic was published by Davis and Cohen in 1996 [8]. The hypothesis is that complete marginal excision with minimal trauma to the nail bed results in the fewest recurrences and optimal cosmesis.

We therefore sought to do a systematic review of the literature to address the following questions: (1) What is the best surgical approach for excising these lesions? (2) What is the age range, sex distribution, and presenting symptoms of subungual exostoses and which toe is most frequently affected? (3) What complications arise from treatment?

Search Strategy and Criteria

We performed the literature search in Medline (1950–May 2013), Cochrane EBM database, and EMBASE (1980–May 2013) provided by OVID; ACP Journal Club (2003–May 2013); CINAHL by EBSCO (1937–May 2013); and PubMed by NLM (1940–May 2013). We used the search terms "subungual exostoses/exostosis", "toe exostoses/exostosis", and "subungual osteochondroma", and we limited our search to human populations. The review was limited to English, French, and Spanish languages. Two authors (MPD, SKG) independently reviewed the abstracts according to predetermined inclusion criteria. Disagreement about inclusion was resolved by consensus with input from the senior author (FFB).

We included studies of diverse designs that reported treatment outcomes for toe exostoses, which comprised 10 or more patients, and excluded case reports and small case series. Two authors (MPD, SKG) independently reviewed the full-text articles (and their respective references) for those that met our inclusion criteria. We graded the articles according to levels of evidence as depicted by Wright et al. [39, 40] and used PRISMA (Preferred Reporting Items for Systematic reviews and Meta-Analyses) guidelines for relevance as determined by Liberati et al. [22].

One of the authors (MPD) collected demographic and treatment data and collated in an Excel spreadsheet (Microsoft Inc, Redmond, WA, USA). All authors contributed to final data analysis and writing of the article.

One hundred twenty-four abstracts were initially identified and screened. Eight articles were excluded because they addressed only upper extremity exostoses or osteochondromas. The remaining 116 full-text articles were reviewed and 13 studies met our inclusion criteria, which were all Level IV retrospective case series comprising 10 or more patients. No experimental or quasiexperimental studies were identified (Fig. 3).

The total number of reported cases in the 13 included studies was 287. A wide variety of medical journals reported on subungual exostosis, suggesting that physicians of diverse specialties have managed this condition. Dermatology journals published 25% of all articles followed by orthopaedics (19%), foot/ankle (13%), and others (44%). Eleven (85%) articles were published in English (Table 1).

Results

Specific operative and nonoperative strategies varied widely and no article compared one technique with another. Histological characterization was mentioned in most of the articles but details regarding tissue preparation, fixation techniques, and staining methods were only included in one [10]. The most successful principles involved curettage, burr, or ronguer débridement down to the base of the stalk to avoid recurrence, with preservation of the nail bed when possible.

Fifty-five percent of the patients were younger than 18 years of age. The female:male ratio was approximately equal (146:143). The average age at the time of presentation was 25.7 years. The hallux was the most common location of the exostoses (204 of 255 [80%]) followed by the second toe (16 of 255 [6%]), third toe (17 of 255 [7%]), fourth toe (12 of 255 [5%]), and the fifth toe (six of 255 [2%]) (Fig. 4). Pain was the most common complaint (193 of 250 [77%]) followed by mass/swelling in the nail (77 of 250 [31%]), nail change (38 of 250 [15%]), or other
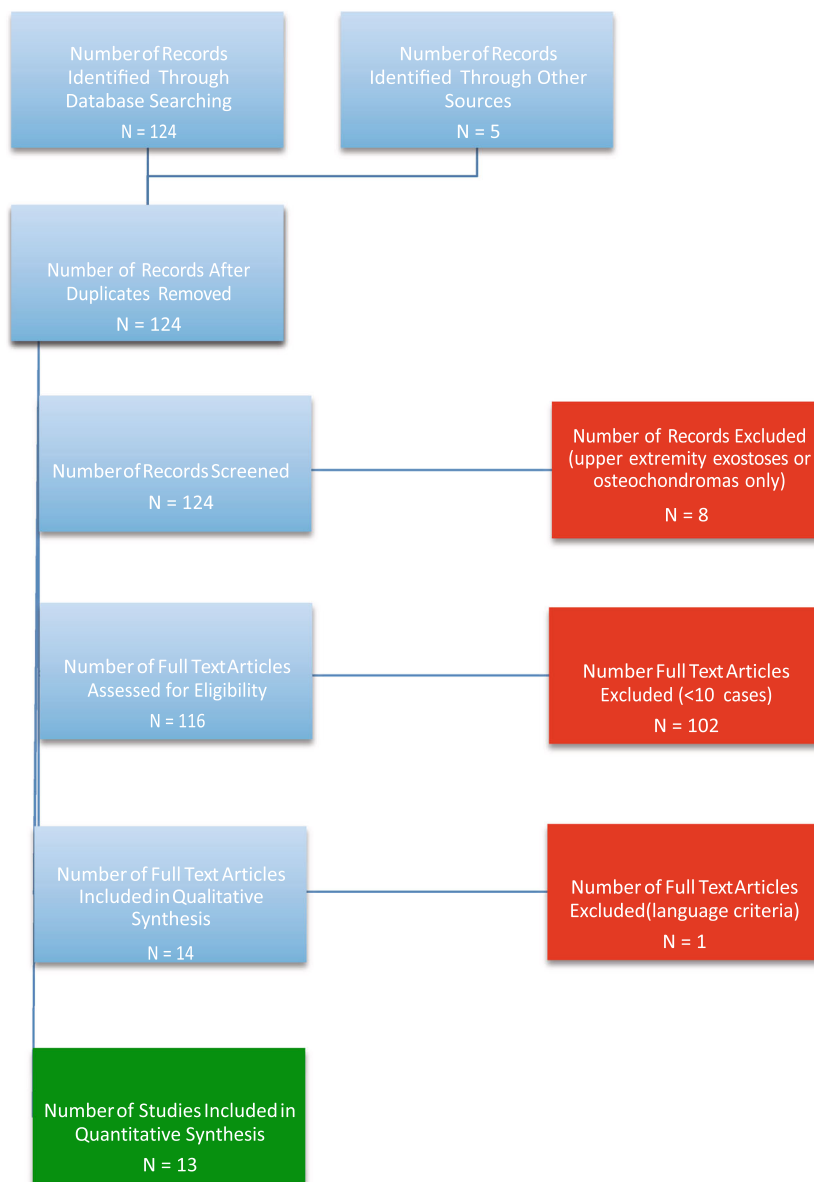
Fig. 3 Flow diagram used to include and exclude relevant articles for the review. Criteria were based on PRISMA guidelines. One hundred twenty-four studies were initially identified through the database screening with 13 meeting all inclusion/exclusion criteria.

complaints such as shoe wear rubbing or stiffness (eight of 250 [3%]) (Table 2). With respect to the history at presentation, the duration of symptoms ranged widely from 2 to 48 months. A history of toe trauma before diagnosis was present in 46 of 160 (29%) documented cases, and a history of previous infection was noted in 21 of 153 (14%) documented cases. There were no reported cases of positive family history or malignant degeneration of the lesion (Table 2). Only those articles reporting antecedent conditions were included in these percentages.

The most prevalent complication was onychodystrophy (malformation or discoloration of the nail), which occurred in 20 of 124 (16.1%) of cases. Strategies aiming to minimize trauma to the nail bed yielded better cosmetic results, mitigating the incidence of onycholysis (loosening or separation of the nail from its bed). A missed, incorrect, or delayed diagnosis occurred in three of 124 (2%) of cases. For all treatments reviewed, recurrence occurred in five of 124 (4.0%) cases, and postoperative infections occurred in four of 124 (3.2%). Other complications such as chronic regional pain syndrome occurred in one of 124 (0.8%) (Table 2). Not all articles reported complications and only those reporting them specifically were included in these percentages.

Table 1. Studies included: general summary

Study	Year	Language	Study design	Level of Evidence	Journal type	Total cases	Pediatric cases	Average age (years)	Male	Female
De Berker and Langtry [9]	1999	English	Retrospective case series	IV	Dermatology	19	9	21	9	10
De Palma et al. [10]	1996	English	Retrospective case series	IV	Foot/ankle	11	2	28.9	5	6
Evison and Price [12]	1966	English	Retrospective case series	IV	Radiology	18	7	24	4	14
Fikry et al. [13]	1998	French	Retrospective case series	IV	Orthopaedic	28	14	18	20	8
Ippolito et al. [16]	1987	English	Retrospective case series	IV	Orthopaedic	27	NR	18.5	10	17
Landon et al. [18]	1979	English	Retrospective case series	IV	Orthopaedic	39	NR	23.5	27	12
Letts et al. [20]	1998	English	Retrospective case series	IV	Trauma	21	21	12.5	10	11
Li et al. [21]	1991	English	Retrospective case series	IV	General	20	9	22	8	14
Miller-Breslow Dorfman [26]	1988	English	Retrospective case series	IV	Pathology	14	9	24.6	7	7
Oliveira et al. [29]	1980	English	Retrospective case series	IV	Dermatology	45	NR	36.5	22	23
Ortega and Sanchez [30]	1988	Spanish	Retrospective case series	IV	Dermatology	20	9	31	5	15
Suga and Mukouda [34]	2005	English	Retrospective case series	IV	Plastics	14	10	25.1	7	7
Valinsky et al. [37]	1991	English	Retrospective case series	IV	Podiatry	11	NR	49	9	2

NR = not reported.

Discussion

Subungual exostoses are relatively uncommon in both the general and pediatric orthopaedic populations. Currently there is a paucity of evidence to define the demographics of this group and to guide management. Most reports on this topic are case series and the most recent attempt at a systematic review is more than 15 years old [8]. The current study extends the scope of that review by including numerous cases reported on since that time and providing a more robust consensus of both the complication profile and successful surgical strategies. This systematic review sought to identify the best treatment approaches, the demographics and common presentations of subungual exostosis, and the complications arising from treatment.

The main limitation of this review is that it is based on a series of Level IV studies, each containing different treatment methodologies and result parameters. This is the result of the fact that the condition is uncommon; thus, designing experimental research comparing different treatment options is a challenge. The synthesis of current literature provided here does, however, establish the
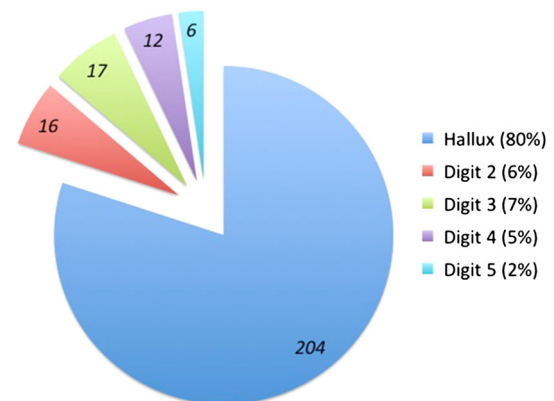

Fig. 4 Pattern of distribution in subungual exostosis of the foot. Total number and percentage from each digit are included.

demographics and optimal treatment strategies on this topic, which is relatively underrepresented in the orthopaedic literature.

We found from our review that the principle of treatment is to achieve complete excision of the lesion by curetting or burring down to normal trabecular bone while

Table 2. Studies included: clinical summary

Study	Predominant complaint	Duration of symptoms	History of trauma	History of infection	Family history	Treatment	Recurrence	Complications
De Berker and Langtry [9]	NR	4–72 months	0	0	0	Excision/secondary intention	Yes x 2	Distal onycholysis in 4 patients
De Palma et al. [10]	Pain	4–32 months	NR	NR	NR	Excision	No	Partial onychodystrophy in 2 patients
Evison and Price [12]	NR	1–18 months	2/18	0	0	Excision	Yes x 4	Infection in 4 patients
Fikry et al. [13]	Pain		21/28	0	0	Excision	NR	NR
Ippolito et al. [16]	Rest pain and walking pain	1–48 months	0	NR	NR	Excision	No	Nail failed to regrow in 8 patients
Landon et al. [18]	Pain	12 months	9/39	15/39	0	Excision, amputation in 1 patient	Yes x 5	NR
Letts et al. [20]	Pain, enlarging nodule, ulceration	NR	3/21	2/21	0	Excision, amputation in 1 patient	Yes x 3	Ingrown toenail in 2 patients, nail regrowing vertically
Li et al. [21]	Pain, nail deformity	3–84 months	3/20	NR	0	Excision, amputation in 3 patients	Yes x 1	NR
Miller-Breslow and Dorfman [26]	Pain in 5 patients, mass in 14 patients	2 weeks to 10 years	0	0	0	Excision, amputation in 1 patient	Yes x 8	Misdiagnosis in 3 patients, incomplete excision in 5 patients
Oliveira et al. [29]	Mass	NR	NR	NR	NR	Excision with "hammer and chisel"	Yes x 3	NR
Ortega and Sanchez [30]	Pain in 15 patients	NR	4/20	NR	NR	Excision	NR	NR
Suga and Mukouda [34]	Pain	NR	4/14	4/14	NR	Excision, including partial nail bed in 9 patients	No	Onycholysis, ingrown toenail
Valinsky et al. [37]	Pain in all except 1, mass in all	NR	NR	NR	NR	Excision and dexamethasone	No	NR

NR = not reported.

Fig. 5A–B Intraoperative images of a marginal excision of subungual exostosis of the great toe in a 15-year-old girl. (**A**) The lesion invaded the nail bed, which was not salvageable; thus, a direct dorsal exostectomy was performed. (**B**) The osteocartilaginous lesion was marginally resected and the base of the stalk was curetted, ronguered, and burred down creating a "saucer-like defect." The sample was sent for pathological analysis and found to be consistent with subungual exostosis. Reproduced with permission from *BMJ Case Reports* [6].

minimizing deformity to the nail plate (Fig. 5). This approach seemed to lead to the lowest incidence of recurrence. De Berker and Langtry [9] described the use of a ronguer to adequately resect lesional tissue, whereas Davis and Cohen [8] used a curette to create a "saucer-like defect" into normal trabecular bone. Suga and Mukouda [34] introduced the idea that if the nail bed was intact, a fish mouth incision distally with a proximally based flap could be raised lifting the nail bed off of the lesion, thus sparing the nail. If the lesion invaded the nail bed, however, a direct dorsal approach should be used (Fig. 5) because the nail bed is already compromised and the priority should be shifted to complete lesional excision. Lokiec et al. [23] showed in his six-patient series that reasonable results in terms of recurrence and cosmesis can

be achieved with only partial nail resection using a distally based flap, which is reattached. Recurrence was related to incomplete excision in most described cases; however, overly aggressive dissection was thought to lead to onychodsytrophy and poor cosmesis [8, 9, 33, 34], particularly when the lesion invaded the germinal matrix [9]. In the single podiatric article included in the study, Valinsky et al. [37] used local anesthesia, a laterally based incision, and a single application of intralesional corticosteroid and antibiotics. There is no mention, however, of results or complications.

Although multiple specific surgical techniques and postoperative wound closure strategies have been reported [6, 8, 9, 23, 34, 37], as a result of a lack of comparative analyses, no one technique has significant evidence to show its superiority. The principle, however, of a balance between complete lesional excision and delicate separation from underlying nail bed structures is the most important factor in mitigating recurrence and nail problems.

Our review has shown some similarities as well as some differences as compared with previous reviews with respect to the epidemiology of subungual exostoses of the toes. The average age of onset of 26 years and the preponderance for the great toe (80%) corroborate with numbers published previously [8, 33, 34]. Conversely, we found that there is a much higher proportion of cases in the pediatric population (55%), and the female:male ratio is closer to 1:1 (compared with 16% and 2:1, respectively, as reported by Davis and Cohen [8]). Also, this review finds that the association with previous trauma or infection is high (29% and 14%, respectively). Although Fikry et al. [13] showed a relationship to previous trauma in 21 of 28 (75%), the remainder of the studies reporting on this was much lower. Similarly, Landon et al. [18] showed prior infection was observed in 15 of 39 (38%), but multiple others showed very little correlation (Table 2). The differences in our results compared with the next most recent review by Davis and Cohen in 1996 [8] are likely attributable to the fact that five of 13 of the articles that met our comprehensive inclusion criteria have been published since the Davis and Cohen study, representing a more recent summary of the current literature.

The complication profile presented in this review shows that important, potentially avoidable morbidity is associated with the treatment of subungual exostosis, including postsurgical deformity (16%), recurrence (4%), and delayed diagnosis (2%). Although the complication of postsurgical nail deformity is related to the size and location of the lesion at presentation, meticulous surgical technique and wound closure can minimize deformity [9, 19, 33]. Although the actual rate of defined "delayed diagnosis" was found to be only 2%, this number is likely much higher. This can be inferred from the duration of

symptoms ranging in many cases well beyond 6 months and up to many years (Table 2). Delayed diagnosis probably stems from the wide differential diagnosis, the broad spectrum of health practitioners from whom patients first seek care, the requirement for appropriate pathological analysis, and the lack of experience of some of those providers in clinically recognizing and managing these relatively rare conditions. Radiographs of the involved toe play an important role in making the differential diagnosis. The recurrence rate likely is a result of incomplete surgical excision. Some authors describe no onycholysis even after they excised a portion of the nail bed through a fish mouth incision and allowed the wound to close secondarily. This seems counterintuitive but may represent the fact that the healing potential is greater in skeletally immature individuals as long as the germinal matrix is maintained. Suga and Mukouda [34] suggest that in the case in which the lesion creates a significant nail bed defect, you can either allow the defect to heal spontaneously or augment the wound with artificial skin and allow it to epithelialize. Recently, DaCambra et al. [6] suggested a novel method of wound management using a vacuum-assisted closure device to promote faster healing with good results. With this variability of management, onycholysis remains an important complication, which can be mitigated with preservation of the nail bed and germinal matrix.

In conclusion, there is weak evidence to guide management of subungual exostosis, a condition that has a relatively high complication rate and which affects children and adults. This topic is underrepresented in the orthopaedic literature, because many of the important clinical series have been published in journals from other specialties [4, 8, 9, 12, 15, 20, 21, 29, 30, 33, 34, 41], reflecting the fact that the condition is treated by many kinds of nonorthopaedic providers. Complete marginal excision of the exostosis appears to mitigate recurrence. Adequate wound management postexcision aiming to minimize disruption to the nail bed and matrix may prevent onychodystrophy, which is a common complication of treatment. Further empirical research is required to compare the outcomes of diverse strategies for wound management postexostosis resection.

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